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Estimating the Effectiveness of Screening for Scoliosis: A Case-Control Study

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ABSTRACT

OBJECTIVE. The aim of this study was to test the hypothesis that screening for scoliosis is effective in reducing the need for surgical treatment.

METHODS. The study was a case-control study. A total of 125 consecutive patients who were treated surgically for idiopathic scoliosis between January 2001 and October 2004 and who were born on or after January 1, 1984, were invited; 108 agreed to participate. A total of 216 control subjects were selected randomly and anonymously, matched with respect to age and gender. For 279 adolescents, exact screening exposure and outcomes could be analyzed. Case subjects were recruited from 4 university and 6 nonuniversity Dutch hospitals; control subjects were recruited from all 37 municipal health services in the Netherlands.

RESULTS. Screen-detected patients received diagnoses at a significantly younger age than did otherwise-detected patients (10.8 ± 2.6 vs 13.4 ± 1.7 years). In total, 32.8% of the surgically treated patients had been screened between 11 and 14 years of age, compared with 43.4% of the control subjects. The odds ratio for being exposed to screening was 0.64. In total, 28% of the patients were diagnosed as having scoliosis before 11 years of age.

CONCLUSIONS. Our results showed no evidence that screening for scoliosis reduced the need for surgery. Abolishing screening seems justified, especially because the effectiveness of early treatment with bracing is still strongly debated. A randomized, controlled trial on the effectiveness of treating patients with idiopathic scoliosis with bracing is urgently needed.

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Key Words

scoliosis, screening, case-control study, effectiveness

Abbreviations

IS—idiopathic scoliosis
RCT—randomized, controlled trial
OR—odds ratio
CI—confidence interval
MHS—municipal health service

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IDIOPATHIC SCOLIOSIS (IS) is defined as lateral curvature of the spine (minimal Cobb angle of 10°), of unknown origin, with concomitant vertebral rotation.¹ Screening for IS was introduced in the United States and many other countries in the 1970s.² Screening aims at detecting patients in an early stage of the clinical course, to allow brace treatment with the aim of preventing further progression and the need for surgical treatment.³ In the Netherlands, an estimated 80% of children are screened for IS at least once.

To date, however, the effectiveness of such screening (and early treatment) has not been established sufficiently, because of a lack of randomized, controlled trials (RCTs).⁴ Some studies concluded that screening for scoliosis is effective,^{2,5-7} whereas others doubted the effectiveness or even considered such screening to be unethical.⁸⁻¹⁰ The US Preventive Services Task Force recommends against the routine screening of asymptomatic adolescents for IS, because evidence has shown that the balance of benefits (a few) and harms (more than a few) is negative.¹¹ Another important issue is that, because the cause of this form of scoliosis is unknown, the current screening test and the early treatment may not be the most appropriate ones.

Earlier, we found that screen-detected patients were detected at an earlier stage of the clinical course and that screen-identified patients had a 73% lower chance of needing surgery.¹² However, overtreatment bias and

length-biased sampling, leading to overestimation of the effect of screening, could be serious problems in such a study.^{12,13}

The present study investigated the effect of screening for scoliosis on reduction of the need for surgery. From a methodologic viewpoint, a RCT is the best design to establish the effectiveness of screening for scoliosis. Because IS is not a common condition, however, such a design would require a very large study population to gain sufficient power, which makes a RCT less feasible.¹⁴ The second-best design is a case-control study,¹⁵ which we performed with individual data on exposure to screening.

METHODS

Design

This was a case-control study, in which the case group consisted of patients with IS who were treated surgically and the control group consisted of a random sample of Dutch youths. Control subjects were matched to case subjects with respect to age and gender. Matching with respect to age was performed to provide equal opportunities for case subjects and control subjects to be exposed to screening in the past. Because IS occurs more often in girls than in boys, matching with respect to gender was applied.

Study Population

Case Subjects

In the Netherlands, ~50 patients with IS each year need surgical treatment; >90% of these operations take place in 11 hospitals. Orthopedic surgeons in 10 hospitals (4 university hospitals and 6 nonuniversity hospitals) in the Netherlands where patients with IS are treated conservatively and surgically were willing to cooperate in this study. They were requested to report all patients who were treated surgically for IS between January 2001 and October 2004 and who were born on or after January 1, 1984. These patients ($N = 125$) were invited to participate in this study; 108 patients (86%) and their parents (if the patient was <16 years of age) gave informed consent for participation.

Control Subjects

The control subjects were matched to the case subjects with respect to age and gender; all control subjects were selected randomly from the source population. For each case subject, 2 control subjects were sampled. Data on the control subjects were collected from the databases of all municipal health services (MHSs) in the Netherlands ($N = 37$), which include almost all youths. Each time a case subject was included in the study, we selected randomly 2 MHSs, weighted with respect to the number of youths registered in each MHS. We requested the MHS to select a control subject whose date of birth and gender matched those of the case subject. Because it was possible that a MHS would have >1 match in its database, we requested that the MHS start searching family names from a certain letter of the alphabet until the first match

was found; these letters were distributed randomly among the control subjects. If there was no match in the database, then the MHS was requested to search for a control subject who was born 1 day after the case subject. If that still did not lead to a match, then the MHS had to search for a control subject born 1 day before the case subject and then, if necessary, 2 days after the case subject was born, and so forth, until a match was found. Such adjustments needed to be made for only 9 control subjects (the maximal difference that was needed was 5 days). We emphasized that the gender of the control subject had to be the same as that of the case subject. Because the control subjects were kept anonymous for the researchers, informed consent from the control subjects was not necessary. The study was conducted according to the principles of the Declaration of Helsinki. Under Dutch law, observational health surveys are exempt from needing approval from a medical ethics committee.

Variables and Measurements

Case Subjects and Control Subjects

School physicians received a questionnaire that they were requested to complete. Data on being exposed to screening, ages at screenings, and whether Adam's forward bending test was performed were collected from youth health care files. Being exposed to screening was defined as being examined for scoliosis during a periodic medical examination or a "single scoliosis screening" with at least Adam's forward bending test.³ All examinations that were performed at the specific request of someone (for example, a gym teacher or parent) and that were not incorporated into the usual screening program were not considered exposure to screening.

Case Subjects

Data on the Cobb angle, curve type, brace treatment, age at detection otherwise (ie, not through screening), and age at diagnosis were collected by using medical files and telephone interviews with participating patients being treated for scoliosis. If the school physician had detected the scoliosis, then the date of detection was retrieved from the youth health care file. The orthopedic surgeon established the diagnosis of IS.

Statistical Analyses

Power Calculations

In total, 102 case subjects and 204 control subjects were needed to obtain a probability of 80% for establishing a 50% reduction in surgery for patients with IS, with $\alpha = .05$.

Statistics

Because some data were skewed and some subgroups were smaller than $n = 30$, we used the Mann-Whitney U test to evaluate significant differences between screen-detected and otherwise-detected patients with respect to median Cobb angles, age at detection, age at diagnosis,

TABLE 1 Characteristics of the Scoliosis Patients Treated Surgically

	Total Group (n = 107)	Screen-Detected (n = 43) ^a	Otherwise-Detected (n = 57) ^a
Gender, n (%)			
Girls	86 (80)	35 (81)	45 (79)
Boys	21 (20)	8 (19)	12 (21)
Curve type, n (%) ^b			
Thoracic	29 (31)	14 (36)	14 (29)
Thoracolumbar	21 (23)	11 (29)	10 (20)
Lumbar	1 (1)	1 (3)	0 (0)
Double thoracic	3 (3)	1 (3)	1 (2)
Double thoracic lumbar	39 (42)	11 (29) ^c	24 (49) ^c
Cobb angle, mean \pm SD (median)			
At diagnosis ^d	42 \pm 15.8 (43)	34 \pm 16.1 (35) ^e	46 \pm 13.3 (46) ^e
Before surgery ^f	56 \pm 10.4 (55)	54 \pm 8.2 (53)	57 \pm 11.7 (56)
After surgery ^g	32 \pm 11.3 (30)	30 \pm 12.9 (28)	33 \pm 10.2 (34)
Duration of brace treatment before surgery, mean \pm SD (median) (n = 46), y ^h	2.5 \pm 2.1 (2) (n = 46)	2.9 \pm 2.4 (2) (n = 25) ⁱ	1.9 \pm 1.6 (1.4) (n = 18) ⁱ

^a For 7 patients, data on being screen detected or otherwise detected were missing. For the screen-detected patients, the school physician clearly exerted the most influence in the screening and/or referral process.

^b For 14 patients, the exact curve type could not be clearly extracted from the medical files.

^c $P < .05$.

^d Missing n = 17.

^e $P < .01$.

^f Missing n = 44.

^g Missing n = 44.

^h Forty-nine patients were treated with a brace for >6 months before surgery; for 3 patients, the total brace period was unknown (patients who were not treated with a brace were excluded from calculation of the average brace time).

ⁱ For 3 patients, data on being screen detected or otherwise detected are missing.

time period between detection and diagnosis, and age at surgery.

Only screenings that were performed before IS was diagnosed by an orthopedic surgeon counted as exposure to screening. For the control subjects, only screenings that were performed before IS was diagnosed for the matched case subjects were valid.¹⁶ The odds ratios (ORs) and their 95% confidence intervals (CIs) for being exposed to screening were calculated by using binary logistic regression analysis. To account for matching with respect to gender, gender was added as a categorical covariate. We did not use matched analysis (eg, conditional logistic regression analysis) because the matching factors did not influence the exposure measure such that they would lead to bias.

First, the OR for being ever/never screened before diagnosis was calculated. We then calculated the OR for being screened or not between 11 and 14 years of age (ie, the ages at which screening is recommended in the Netherlands). The latter was performed only for the case subjects and matched control subjects who were still eligible for screening (ie, scoliosis had not yet been diagnosed).

Estimation of Costs

Costs for screening for scoliosis were estimated on the basis of the cost of activities model for a MHS.⁴ This model considers that 42% of the screenings are part of a school physician's consultation, 20% a single screening performed by school physicians, 31% part of a school nurse's consultation, and 7% a single screening per-

formed by school nurses. Estimations of the costs of IS surgery were based on Dutch health care fees.

RESULTS

For 7 selected case subjects and 1 control subject, the youth health care files were not retrievable. For 1 selected case subject and 2 control subjects, we did not receive a completed questionnaire; for another 5 case subjects, the age of diagnosis was missing. One case was diagnosed before the age of 5 years. Because this case might have represented some sort of very early-onset scoliosis, we deleted this case subject and the matched control subjects from all analyses.

Table 1 shows the characteristics of the patients with IS that were treated surgically. The ratio of girls to boys was 4:1. Screen-detected patients had significantly smaller Cobb angles at diagnosis, compared with otherwise-detected patients ($P < .01$). After surgery, Cobb angles did not differ significantly between screen-detected and otherwise-detected patients. Approximately one half of the patients were treated with a brace before surgery. Although there was no significant difference in duration of brace treatment between screen-detected and otherwise-detected patients, screen-detected patients had an almost threefold greater chance of being treated with a brace before surgery (OR: 3.1; 95% CI: 1.3–7.0), compared with otherwise-detected patients. On average, brace treatment lasted for 2.5 years.

Table 2 shows mean and median ages at detection, diagnosis, and surgery and the time period between detection and diagnosis. Screen-detected patients were

TABLE 2 Ages at Detection, Diagnosis, and Surgery and Time Between Detection and Diagnosis

	Mean \pm SD	Median
Age at detection, y		
Total	11.1 \pm 2.7	11.4
Screen-detected	9.7 \pm 2.6	10.4 ^a
Otherwise-detected	12.9 \pm 1.6	13.2 ^a
Age at diagnosis, y		
Total	12.0 \pm 2.6	12.4
Screen-detected	10.8 \pm 2.6	11.2 ^a
Otherwise-detected	13.4 \pm 1.7	13.8 ^a
Age at surgery, y		
Total	14.9 \pm 1.6	14.9
Screen-detected	14.7 \pm 1.4	14.8
Otherwise-detected	15.1 \pm 1.8	15.1
Time between detection and diagnosis, y		
Total	0.9 \pm 1.2	0.3
Screen-detected	1.1 \pm 1.4	0.3
Otherwise-detected	0.5 \pm 0.7	0.2

Only patients with available data on both age at detection and age at diagnosis were included in this analysis. In this group ($n = 66$), there were 37 screen-detected patients and 29 otherwise-detected patients. For all 66 patients, data on the age at surgery were available.

^a $P < .01$.

~2.5 years younger at detection (Fig 1) and at diagnosis. On average, there were >12 months between detection and diagnosis for screen-detected patients and 6 months for otherwise-detected patients. In total, 55% of the screen-detected patients and 66% of the otherwise-detected patients were diagnosed by an orthopedic surgeon within 4 months after detection. In Fig 1, the 3 peaks in the screen-detected curve correspond to the ages at which school physicians perform periodic medical examinations in the Netherlands.

The majority (74%) of children had been exposed to screening at least once (Table 3). The proportion of case subjects exposed to screening was slightly greater (80.5%) than the proportion of control subjects exposed to screening (74.0%). The OR for being exposed to screening was 1.44 (95% CI: 0.77–2.68; $P = .25$).

The OR for being screened at the age of 11, 12, 13, or 14 years was 0.64 (95% CI: 0.34–1.19; $P = .16$) (Table 4). The proportion of case subjects exposed to screening was smaller than the proportion of control subjects exposed to screening, but screening did not reduce the chance of surgery significantly. It should be noted that

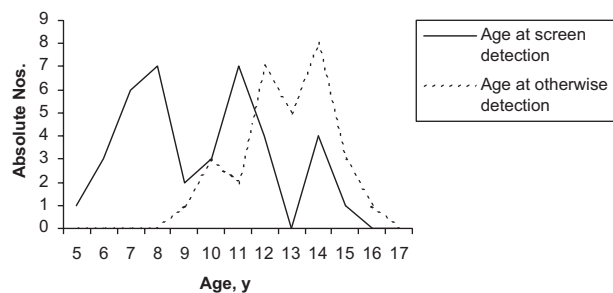


FIGURE 1

Age at screen detection versus age at otherwise detection (data were missing for 39 patients).

TABLE 3 Numbers of Case Subjects and Control Subjects and Their Exposure to Screening

	n (%)		
	Case Subjects	Control Subjects	Total ^a
Exposed to screening	70 (80.5)	142 (74.0)	212 (76.0)
Not exposed to screening	17 (19.5)	50 (26.0)	67 (24.0)

The OR was 1.44 (95% CI: 0.77–2.68; $P = .25$).

^a Because some data on whether the bending test was performed and whether the test could be defined as screening were missing, the numbers do not add up to 107 case subjects and 214 control subjects.

30 case subjects (28%) were diagnosed as having IS before they were 11 years of age.

Costs to screen 80% of 1 Dutch birth cohort (~200 000) were estimated at 3 million euros. Costs for 1 scoliosis operation were estimated at 11 000 euros. Total surgical costs were estimated at 550 000 euros, given 50 operations in 1 year.

DISCUSSION

The results of this study did not show a significant reduction in the need for scoliosis surgery attributable to screening. Patients detected through screening were significantly younger at diagnosis than were patients who were detected otherwise. This means that the screen-detected patients had additional years of concern about the disease and they had a greater chance of brace treatment but without better final outcomes. With detailed data for 200 case subjects and control subjects, we had 80% power to show a 59% reduction in scoliosis operations. Our results confirm the conclusion of Wieggersma et al,¹⁰ who also reported that screening for scoliosis did not reduce the need for surgery.

Case-control studies are susceptible to different kinds of bias. In this study, all patients already suffered from the serious outcome that screening is supposed to prevent; therefore, ascertainment bias is not a serious problem. Because the control subjects were kept anonymous to the researchers, there was no nonresponse in the control group that could lead to nonresponse bias. The response rate for participation among the patients was high (86%). If we had considered the nonresponders as not being exposed to screening, then we would have found a positive effect of screening. However, this is a highly unlikely scenario, because we have no indications that the nonresponders would be different from the responders. Furthermore, being exposed to screening for

TABLE 4 Numbers of Case Subjects and Control Subjects and Their Exposure to Screening at 11 to 14 Years of Age

	n (%)		
	Case Subjects	Control Subjects	Total ^a
Exposed to screening	21 (32.8)	59 (43.4)	80 (40.0)
Not exposed to screening	43 (67.2)	77 (56.6)	120 (60.0)

The OR was 0.64 (95% CI: 0.34–1.19; $P = .16$).

^a Thirty case subjects were diagnosed as having IS before the age of 11 years.

scoliosis depends more on whether the youth health care department offers the screening for scoliosis in general than on the characteristics of the child or the parents; therefore, we do not expect selection bias to influence the results substantially. Recall bias could be a problem for otherwise-detected case subjects in relation to data on the detection date; patients might have underestimated the time between detection and diagnosis. This could also apply to the age at diagnosis if the medical chart was incomplete.

Our results do not show a significant reduction in the need for surgery attributable to screening for scoliosis among children 11 to 14 years of age (the ages at which screening is usually recommended). If we assume that the OR of 0.64 is the true size of the effect, then the costs of keeping 1 patient from the need for surgery are estimated at (at least) 130 000 euros, and ~5800 children would need to be screened. These are relatively high costs and involve considerable effort, given that severe scoliosis is neither common nor fatal. Furthermore, screening identifies some children who ultimately receive treatment but involves referral of many more who do not.¹⁷ Therefore, these costs are an underestimation of the real costs, because they exclude the costs of visits to general practitioners and orthopedic surgeons and of radiographs attributable to false-positive results. Yawn and Yawn¹⁸ calculated that case finding costs for screening were \$10 836 per child treated (conservatively or surgically) for scoliosis.

In our previous study, we found that 2 prerequisites for an effective screening program were met, that is, earlier detection and less surgery in the screen-detected group.¹² However, overtreatment bias and length-biased sampling could not be ruled out. In the present study, we also found that screen-detected patients were diagnosed in an earlier stage and had a greater chance of being treated with a brace. However, we could not prove that exposure to screening led to less surgery. One explanation for this could be that screening for scoliosis may lead to overtreatment with a brace.¹¹ Patients with a relatively small Cobb angle are more likely to be detected through screening than otherwise (eg, by themselves or by their parents). Some of these patients are treated with a brace, whereas they would not have visited an orthopedic surgeon and received or needed treatment at all if they had not been identified through screening.

The relatively low sensitivity (55%) of the screening program¹² could perhaps explain why we did not find a beneficial effect. Furthermore, low levels of compliance with brace treatment could lead to ineffective treatment with a brace, which could result in more operations. More importantly, it is still unclear whether early intervention with a brace is an effective strategy in preventing surgery for patients with IS. Some authors consider a brace effective,^{19,20} whereas others conclude that the effectiveness of bracing is doubtful or they recommend a RCT on brace treatment.^{4,8,21,22} If we had found convincing evidence for the beneficial effects of screening for scoliosis, then this would have implied that early treatment with a brace is effective. Because we did not find

convincing evidence that screening is effective, we need to determine whether early bracing is effective by means of a RCT; such a trial started in the Netherlands in 2006.

CONCLUSIONS

We think that abolishing screening for scoliosis seems justified, because of the lack of evidence that screening and/or early treatment with bracing is beneficial. For now, instead of screening large numbers of asymptomatic children, the appropriate approach would be to look at a child's back when there are indications that something is wrong. Such children should be examined and, if necessary, referred to a specialist. If a RCT on brace treatment establishes that bracing is effective, then it will be worthwhile to determine which children could benefit from a screening program.

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